Case report

Atypical presentation of erythema induratum of Bazin

Apresentação atípica do eritema indurado de Bazin

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ABSTRACT

Cutaneous involvement in tuberculosis is uncommon and includes several clinical entities, including the erythema induratum of Bazin, whose classic presentation involves purplish nodules or nodular plaques that tend to ulcerate and evolve into outbreaks. In this report, we describe a case of atypical clinical presentation of erythema induratum of Bazin in a 17-year-old patient who presented with crusty, scaly and hyperemic lesions on her legs, which had been progressing for approximately 3 years without signs of systemic involvement. Diagnostic investigation corroborated this hypothesis through the epidemiological link and compatible anatomopathological studies and by ruling out the most common differential diagnoses. The patient showed clinical improvement after starting the anti-tuberculosis medications, and the lesions completely disappeared at the end of the treatment.

Headings: Tuberculosis; Extrapulmonary; Tuberculosis; Cutaneous; Panniculitis; Case Reports.

RESUMO

O acometimento cutâneo pela tuberculose é incomum e compreende diversas entidades clínicas, dentre elas o eritema indurado de Bazin, cuja apresentação clássica envolve nódulos violáceos ou placas nodulares, com tendência à ulceração e com evolução em "surtos". Neste relato descrevemos um caso de apresentação clínica atípica do eritema indurado de Bazin numa paciente de 17 anos que apresentava lesões crostosas, descamativas e hiperemiadas em membros inferiores, com evolução progressiva há cerca de três anos e sem quaisquer sinais de comprometimento sistêmico. A investigação diagnóstica corroborou a hipótese aventada através de vínculo epidemiológico e estudo anatomopatológico compatível, bem como por descartar os diagnósticos diferenciais mais comuns. A paciente evoluiu com melhora clínica após o início do tratamento da tuberculose e obteve involução completa das lesões ao final do esquema terapêutico específico.

Descritores: Tuberculose Extrapulmonar; Tuberculose Cutânea; Paniculite; Relatos de Casos.

INTRODUCTION

Cutaneous involvement in tuberculosis is uncommon, corresponding to approximately 1%–2% of all extrapulmonary forms[1-3]. This relatively low occurrence of cutaneous tuberculosis adds to the low positivity of microbiological tests[1], highlighting the importance of clinical presentation that indicate the correct diagnosis together with other direct and indirect tools.

Here, we describe a challenging case of an atypical presentation of cutaneous tuberculosis known as erythema induratum of Bazin.

CASE REPORT

The patient was a previously healthy 17-year-old girl from Guarulhos, São Paulo. She was referred to a reference outpatient clinic for lesions on her legs that presented approximately 3 years before the initial consultation and had worsened in the last 3 months. She reported having previously received treatment with topical corticosteroids prescribed in other services, which, however, had no effect whatsoever. She denied fever, weight loss, sweating, respiratory symptoms, and other systemic problems as well as no previous trauma at the site of the lesions. She also denied gardening- or water-related activities, and had always lived in an urban area and worked administrative jobs. However, she mentioned having contact with a relative diagnosed with pulmonary tuberculosis approximately 14 years ago.

On physical examination, she had crusty, hyperemic, scaly lesions with irregular borders on both legs. These lesions were painless, nonpruritic, with preserved tactile and thermal sensitivity. The largest lesion on the back of the right leg measured 10 cm (Figure 1). According to the patient, the lesions had always had the same appearance but had recently increased in size, especially on the right leg.

In the initial approach, an infectious etiology (fungal or mycobacterial) was deemed most likely; however, the likelihood of an autoimmune disease or cancer could not be rolled out. Laboratory tests, namely blood count, inflammatory markers (C-reactive protein and erythrocyte sedimentation rate), autoantibody testing, kidney function and liver enzymes, were requested to investigate associated systemic diseases, comorbidities, and immunosuppression. However, all results were within normal limits. Additionally, serologies for HIV, syphilis, and hepatitis B and C were nonreactive. Despite the absence of respiratory complaints and given that our patient had contact with a patient diagnosed with tuberculosis, a chest CT scan was warranted but the result was normal. The purified protein derivative (PPD) skin test, however, showed a 19-mm induration, and the interferon-gamma release assay (IGRA) was positive; thus, confirming the diagnosis of latent infection by *Mycobacterium tuberculosis*.

It was then decided to perform a biopsy of the lesion on the right leg. The histological findings showed lobular panniculitis with granulomatous areas, lymphocytic infiltrate, and slight eosinophilia; however, direct fungal examination and bacilloscopy were negative. Another biopsy sample from the left leg revealed fibrosis and angiomatosis with a focal and residual chronic granulomatous inflammatory process, also with negative fungal and bacilloscopy. It was finally decided to start specific treatment for tuberculosis using a basic regimen (rifampicin, isoniazid, pyrazinamide, and ethambutol). Gradual improvement was observed, and the lesions completely disappeared after 6 months at the end of the treatment period, with only residual hyperpigmentation remaining. Lesion recurrence was not observed during the 18 months of follow-up (Figure 2).

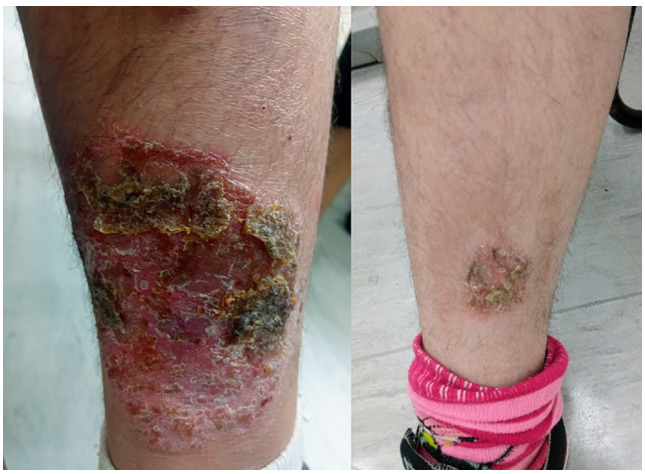

Figure 1. Skin lesions on the posterior (A) and anterior (B) regions of the right leg before tuberculosis treatment.

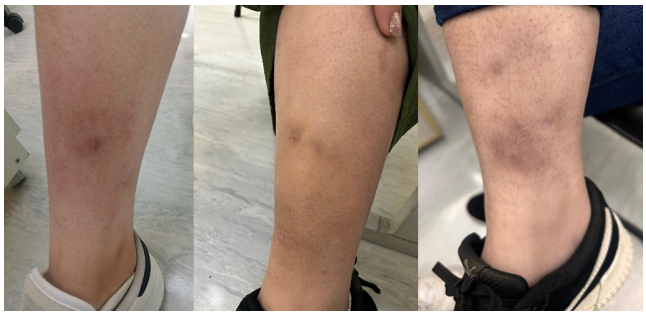

Figure 2. Hyperchromic sequelae spots in the anterior region of the right leg at the end of treatment (A) and in the posterior (B) and anterior (C) regions of the same right lower limb after 18 months of treatment.

DISCUSSION

First described in 1826 by Theophile Laennec (even before the isolation of *M. tuberculosis* by Robert Koch in 1882), cutaneous tuberculosis comprises several clinical entities that vary depending on the immune status and the humoral and cellular response pattern of the host, the route of inoculation of the microorganism into the skin, and bacteriological characteristics like the pathogenicity of the strain[1,2]. Cutaneous tuberculosis can be classified into: (1) exogenous, when there is primary inoculation of the pathogen, e.g., tuberculous chancre and tuberculosis verrucosa cutis; (2) endogenous, when there is hematogenous dissemination through contiguity or autoinoculation, e.g., scrofuloderma, orificial tuberculosis, lupus vulgaris, acute miliary tuberculosis, and gummatous tuberculosis; and (3) tuberculids, when there is a hypersensitivity reaction to *M. tuberculosis,* e.g., nodular vasculitis, papulonecrotic tuberculid, lichen scrofulous, and erythema induratum of Bazin (EIB)[1,4,5].

The diagnosis of EIB is based on clinical presentation, epidemiological history (further evidenced by PPD and IGRA), compatible histological findings, and as in our case, response to treatment[6-8]. Clinically, EIB presents as a nodular eruption with chronic progression, classically described as violaceous nodular plaques with ulceration tendency that usually evolve in outbreaks. The lesions most typically affect the lower limbs and are more common in young women[1,4-6,9]. In the present case, the patient was a women in the most commonly affected age group, and her skin lesions corresponded to the usual topography of EIB. However, they were scaly, crusted, without regular borders with a fixed and gradually worsening pattern, and did not follow the classic violaceous nodular plaques observed in such outbreaks. Consequently, other forms of cutaneous tuberculosis were also considered. However, the patient did not have a history suggestive of possible external inoculation of the pathogen (the manner in which tuberculosis verrucosa cutis is acquired), and there were no signs of active tuberculosis in other sites, including the lungs, making hematogenous dissemination, a characteristic of scrofuloderma, unlikely[1]. Notably, laboratory results did not suggest fungal infection or autoimmune disease, and the biopsies ruled out the possibility of cancer.

Histologically, EIB is characterized by septal, lobular, or mixed granulomatous panniculitis, with an infiltrate comprising lymphocytes, histiocytes, plasma cells, and multinucleated giant cells, while caseous necrosis may also occur[1,4]. A certain degree of vasculitis involving arteries and veins in the adipose tissue may be present but is not considered as a requirement to diagnose EIB[4,6,8,10-12]. However, the negative results of bacilloscopy and DNA research for M. tuberculosis are striking in this entity, despite the mycobacterial culture may eventually be positive in some cases. Conversely, PPD and IGRA are always positive[1,4,9,13,14]; although EIB can occur with active tuberculosis in other sites[8,9]. The histological findings of the biopsy performed on the patient's right leg matched the classic description of EIB in literature, including the absence of vasculitis[4,10,12], which may also explain the absence of necrosis[1,2]. Despite the granulomatous infiltrate, the histology of the left leg sample differed from the classic description owing to the absence of panniculitis, which may be explained by the biopsy site and the long course of the disease[4,12]. Moreover, we highlight a peculiarity of this case, namely the presence of eosinophils in one of the samples, a histological finding undescribed in the literature on EIB[1,4,12]. The hypersensitivity reaction in EIB is mediated by type IV cells with the participation of T lymphocytes, macrophages, and Langerhans cells[1,4,5,7], not by eosinophils, which again denotes the pleomorphic spectrum of presentation of the disease.

With regard to treatment, the use of basic tuberculosis medications achieves a good clinical response, and this was the case with our patient[1,4,5]. Some authors suggest the need to extend the treatment for longer periods, whereas others advocate maintaining monotherapy with isoniazid after completing the basic regimen. Moreover, others still argue in favor of using doxycycline, potassium iodide, dapsone, and thalidomide as adjuvants to control the inflammatory component[1,9].

CONCLUSION

EIB is an uncommon form of tuberculosis and the case reported here is a good example of the occurrence of atypical forms that need to be taken under consideration when addressing chronic skin lesions, even without microbiological, molecular, or histological evidence of mycobacteria in the biopsy samples. The epidemiological history corroborated by the diagnosis of latent tuberculosis was of great importance in this case, and the good response to empirical treatment facilitated specific diagnostic definition.

"This case report deserved an official declaration of acknowledgement and ethical approval by its institution of origin and was peer-reviewed before publication, whilst the authors declare no fundings nor any conflicts of interest concerning this paper. It is noteworthy that case reports provide a valuable learning resource for the scientific community but should not be used in isolation to guide diagnostic or treatment choices in practical care or health policies.

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